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Case Report

Gingival plasma cell granuloma

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ABSTRACT

Plasma cell granuloma, also known as inflammatory pseudotumor is a tumor-like lesion that manifests primarily in the lungs. But it may occur in various other anatomic locations like orbit, head and neck, liver and rarely in the oral cavity. We here report an exceedingly rare case of gingival plasma cell granuloma in a 58 year old woman who presented with upper gingival polypoidal growth. The histopathological examination revealed a mass composed of proliferation of benign spindle mesenchymal cells in a loose myxoid and fibrocollagenous stroma along with dense infiltrate of chronic inflammatory cells predominantly containing plasma cells. Immunohistochemistry for kappa and lambda light chains showed a polyclonal staining pattern confirming a diagnosis of plasma cell granuloma.

Key Words: Inflammatory pseudotumor, plasma cell granuloma, plasma cells, polyclonal plasma cells

INTRODUCTION

Gingival Plasma cell granulomas are extremely rare. These are non-neoplastic, tumor-like lesions of unknown etiology and are composed predominantly of polyclonal plasma cells. In 1968, Bhaskar, Levin and Firch first reported the cases of gingival plasma cell granuloma.[1] Although Plasma cell granuloma (PCG) occurs most commonly in lungs, other organs may be involved. In head and neck, the areas most commonly involved are the orbit and paranasal sinuses, but they have been also described in the larynx, pterygomaxillary space, tonsils, ears, tongue, lip, oral mucosa, periodontal tissues and rarely gingiva.[2] Literature reviewed shows that gingival plasma cell granuloma is exceedingly rare and very few case reports of gingival plasma cell granuloma have been observed. Intraoral PCG occurs in a wide age range of 19 months to 63 years, but most of the cases of gingival PCG are observed in 4thand 5th decades of life and there is a slight female predominance.[3,4] Clinically gingival PCG presents as a nodular, polypoidal mass with smooth surface. It does not produce significant systemic symptoms. Routine laboratory examination is normal and microbiological culture results are negative. Radiologically some oral lesions have shown infiltrative margins giving an appearance of a malignant tumor.[5] Hence such lesions should be histologically examined to decide the exact nature of these lesions.

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CASE REPORT

A 58-year-old woman was admitted in November 2010 who presented with an enlarging, painless mass in the oral cavity. The mass was present since five years and was slowly increasing in size. There was no history of trauma or surgery to the oral cavity. She had no systemic symptoms. On oral examination, the mass was polypoidal, nontender, firm measuring 3 × 2 cms and was located on the inner aspect of upper gingiva extending from right middle incisor to the left canine region. The mass did not involve the palate. Radiological examination and serum

electrophoresis were normal. Routine laboratory examination was normal. The mass was excised and sent for histopathological examination.

Pathologic findings

Grossly, the lesion was polypoidal and solid measuring 3 × 2 × 1.5 cms with smooth white cut surface [Figure 1]. Microscopically, the mass was lined by stratified squamous epithelium with focal ulceration. The mass was composed of nodular infiltrates of mature plasma cells admixed with lymphocytes and histiocytes on the background of loose myxoid and collagenized stroma showing scattered fibroblasts and myofibroblasts [Figures 2 and 3]. In areas, the lymphoplasmacytic infiltrate was prominent around the blood vessels. Russell bodies were also seen [Figure 4]. Mitotic figures or nuclear atypia were not seen. Immunostaining for kappa and lambda light chains revealed a polyclonal plasma cell population [Figures 5 and 6].

DISCUSSION

Plasma cell granuloma is a rare tumor like lesion characterised histologically by fascicles of spindle mesenchymal cells admixed with chronic inflammatory cells predominantly plasma cells. It has various components like fibroblasts, myofibroblasts, inflammatory cells (plasma cells, lymphocytes, histiocytes, mast cells and eosinophils). The stroma is collagenous and/or myxoid. All these components are arranged in varying proportions and thus create a marked histological diversity. Depending upon the predominant components, it has various nomenclatures like plasma cell granuloma, plasma cell pseudotumor, inflammatory pseudotumor, inflammatory myofibroblastic tumor, and myofibrohistiocytic proliferation.[6]

The aetiology of PCG/inflammatory pseudotumor (IPT) is unknown. The histologic diversity has led to conflicting opinions regarding the inflammatory or neoplastic nature of this lesion. The finding of human herpesvirus-8 DNA sequences and over expression of human interleukin 6 and cyclin D1 has been recently reported in seven cases.[7] Kim *et al*. suggested that interleukin-6 (IL-6) and phospholipase C-γ1 may induce heavy plasma cell infiltration in cyclosporine-induced gingival overgrowth.[8] Debate exists about the inflammatory or neoplastic nature of this lesion, with majority of reports siding with the post inflammatory reactive process. Some cases show a predominance of mature plasma cells and lymphocytes that are mixed with histiocytes and only a minor mesenchymal component. The plasma cells are polyclonal[6,7] favouring inflammatory nature. Other cases are composed predominantly of bland fibroblasts and myofibroblast spindle cells arranged in interlacing fascicles or storiform pattern with only a minor component of inflammatory cells. The spindle cells stain positive with antibodies to vimentin and actin, and rarely, occasional cells stain with desmin, which is consistent with fibroblasts and myofibroblasts. However spindle cells of some lesions have been shown to possess a persistent abnormality involving chromosome 2p23 a ALK gene locus, which is consistent with a neoplastic nature of this lesion.[9]

PCG/IPT may be misinterpreted by the pathologists as nodular fasciitis, fibromatosis, fibrosarcoma or plasmacytoma. Nodular fasciitis rarely occurs in the oral cavity and it is characterised histologically by the presence of loose myxoid matrix containing short linear curved fascicles of spindle cells. Fibromatosis of the oral cavity usually occurs in young adults and it is characterised histologically by broad interlacing fascicles of mature fibroblasts with a variable degree of collagenisation. An inflammatory component is absent. Oral PCG needs to be distinguished from the recently described follicular dendritic cell tumor of the oral cavity, which runs an indolent course with a tendency of local recurrence. It can closely mimic inflammatory pseudotumor with whorls or fascicles of plump spindle cells in an inflammatory background of lymphocytes and histiocytes. In contrast, plasma cells constitute a significant proportion of the chronic inflammatory cells in inflammatory pseudotumor. The distinction can be established by the positive staining for CD21, Ber-MAC-DRC, and Ki-M4 in follicular dendritic cell tumor.[3]

In view of predominance of plasma cells in our case the differential diagnosis considered was plasmacytoma. In plasmacytoma, there are diffuse sheets of neoplastic, variably differentiated, monoclonal plasma cells. Mitotic activity and amyloid deposition may be present and the inflammatory cells are very sparse.[10] The present case showed admixture of lymphocytes and plasma cells along with Russell bodies. Immunohistochemistry showed polyclonal plasma cells. The other polyclonal lesions of gingiva include plasma cell gingivitis, which is usually not a

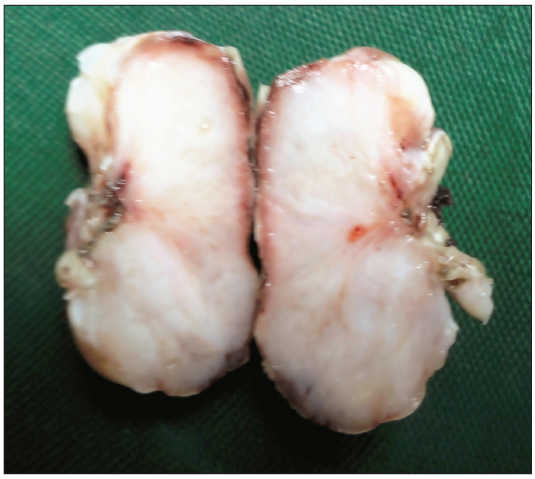

Figure 1: Gross photograph showing polypoidal mass with smooth white cut surface

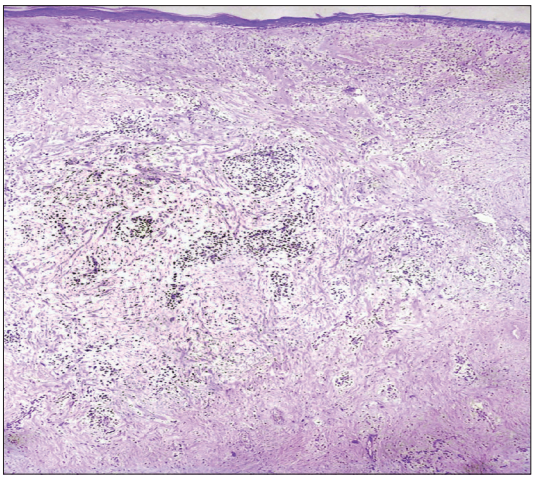

Figure 2: Histological picture showing spindle cell proliferation admixed with dense Lymphoplasmacytic infiltrate (H and E ×100)

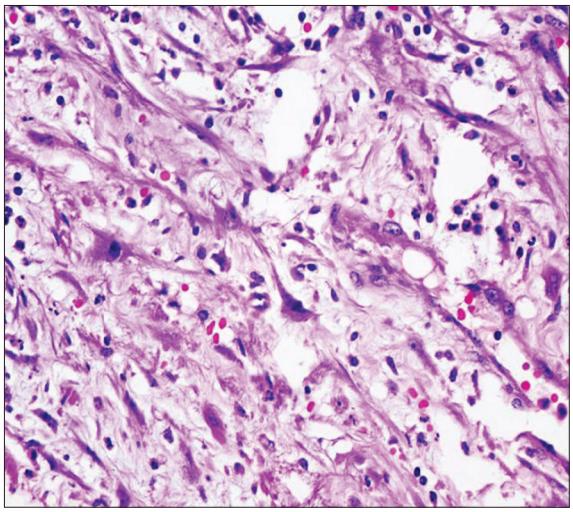

Figure 3: Histological picture showing fibroblasts and myofibroblasts (H and E ×400)

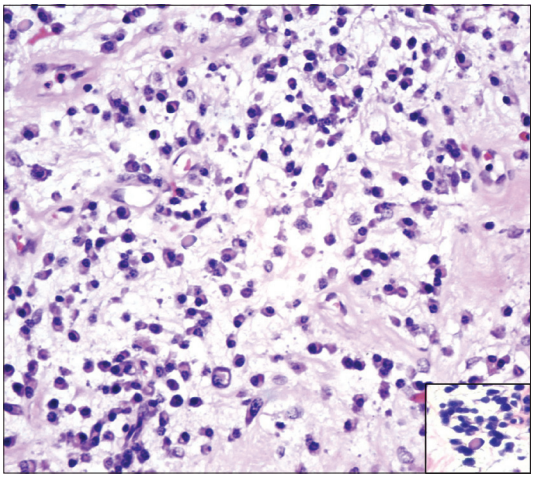

Figure 4: Histological picture showing predominance of plasma cell along with Russell bodies.Inset shows Russell body (H and E ×400)

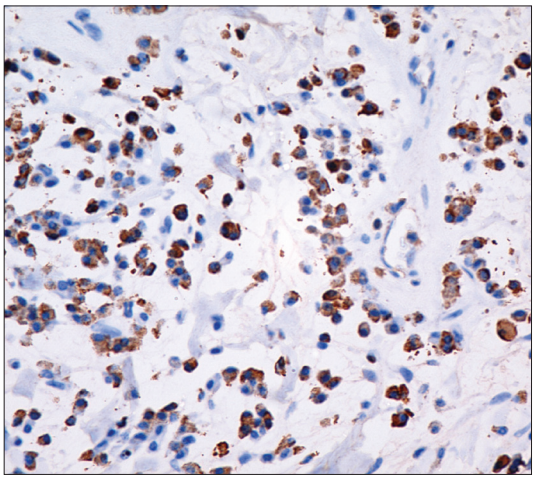

Figure 5: Immunohistochemistry for kappa chains

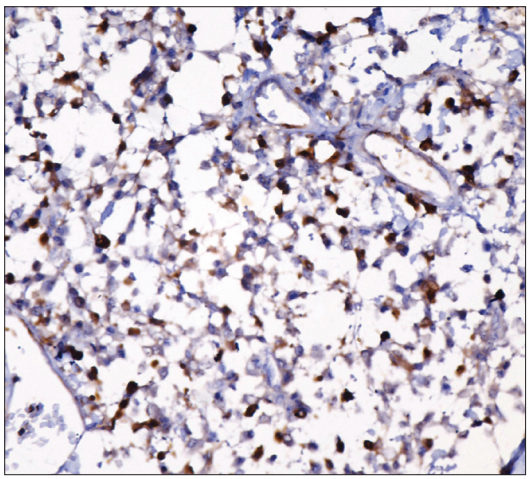

Figure 6: Immunohistochemistry for lambda chains

localised nodular lesion but presents as generalised oedematous and erythematous elevations.[10]

Naderi *et al*. in their study of 2068 cases of reactive lesions of oral cavity found that peripheral giant cell

granuloma was the most prevalent lesion of gingiva.[11] However plasma cell granuloma of gingiva is a very rare lesion. In 1968 Bhaskar, Levin and Firch[1] reported 45 cases of PCG of periodontal tissue and this appears to be the first report on this pathologic entity on the gingival tissue. Thereafter very few case reports have been documented in the literature. Most of these are in the form of single case reports. Acevedo and Buhler,[12] Earl and lowry,[4] Ide and Shimoyama,[4] Peacock ME,[13] Shin JM,[14] Karthikeyan and Pradeep,[10] Baltaciaglu,[1] Namboodiripad,[15] Phadnaik,[1] Balaji Manohar and S Bhuvaneshwari,[16] have described single case reports of PCG of the gingiva. Their clinical presentation and histopathological findings are similar to those observed in the present case. KIM SS and Eom D have described two cases of PCG in cyclosporine induced gingival overgrowth.[8] In January 2011 Kim YS, Lee SK described 14 cases of PCG out of 59 chronic inflammatory gingival lesions examined. They divided the gingival plasma cell granuloma into three histological types viz. plasma cell predominant type (PPT), mixed inflammatory cell type (MICT), and sclerosed fibrosis type (SFT). The results of immunohistochemical studies on these cases suggest that a gingival plasma cell granuloma shows variable gene expression for cell-mediated immunity and stromal tissue degeneration, undergoing sclerotic fibrosis with a persistent inflammatory reaction.[17] Idle *et al*. States that pure PCG should not be called as inflammatory pseudotumor and the term IMT should be applied only for genuine lesions of myofibroblasts.[4]

PCG in the oral cavity is usually benign and simple excision of the lesion is curative. In our case the patient was followed up for 6 months after the surgery. During this period the patient had no recurrence of the lesion. After that the patient was lost for follow up. Although surgery is the principal treatment, regression and response to corticosteroids and nonsteroidal inflammatory agents have been noted in rare cases.[7]

CONCLUSION

Plasma cell granuloma of the gingiva is a rare entity that may be confused with a malignant tumor on clinical and radiographic grounds.[5] The gross and microscopic similarities to other oral spindle cell tumors can also be misinterpreted as those of a more aggressive lesion. So awareness of oral PCG/inflammatory pseudotumor and its distinctive morphologic features is important in avoiding the misdiagnosis. It is also important to recognize this entity as a benign inflammatory lesion to avoid unnecessarily extensive and potentially destructive surgery. We report here this case for its rarity.

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